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METHODS

Estimating non-response bias in family studies: Application to mental health and lifestyle

Jacqueline M. Vink, Gonneke Willemsen, Janine H. Stubbe, Christel M. Middeldorp, Rozemarijn S.L. Ligthart, Kim D. Baas, Hanneke J.C. Dirkzwager, Eco J.C. de Geus & Dorret I. Boomsma

Department of Biological Psychology, Vrije Universiteit, Amsterdam, The Netherlands

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Abstract. Non-response to mailed surveys reduces the effective sample size and may introduce bias. Non-response has been studied by (1) comparison to available data in population based registers, (2) directly contacting non-respondents by telephone or single-item reply cards, and (3) longitudinal repetition of the survey. The goal of this paper was to propose an additional method to study non-response bias: when the variable of interest has a familial component, data from respondents can be used as proxy for the data from their non-responding family members. This approach was used with data on smoking, alcohol consumption, physical activity, coffee- and tea-use, education, body mass index, religion, burnout, life events, personality and mental health in large number of siblings and DZ twins registered with the Netherlands Twin Register. In addition, for smoking

behavior, we also used the second strategy by sending a reply card. Results show that scores of members from less cooperative families or incomplete twin pairs tended to be more unfavorable than the scores from highly cooperative families or complete twin pairs. For example, family members from less cooperative families cycled less often and scored higher on anxious depression and neuroticism. For smoking, both the results of the reply card and the results of the additional method suggested a higher percentage smokers among the non-respondents but this was only significant with reply card method. In general, differences between highly/less cooperative families and complete/incomplete DZ twins were small. Results suggest that, even for studies with moderate response rates, data collected on health, personality and lifestyle are relatively unbiased.

Key words: Health and lifestyle, Mailed survey, Mental state, Non-response, Personality, Twin-family design

Introduction

Mailed surveys are widely used to collect data on health and lifestyle in large populations. In Europe, response rates to mailed surveys vary from 52 to 95% [1]. Non-response to mailed questionnaires reduces the effective sample size and therefore the statistical power of the study. Moreover, survey results will be biased by non-participation if refusal to participate is not distributed randomly, and is either directly or indirectly related to the traits under study. Although studies usually recognize the risk of response bias, they are often unable to quantify the degree of bias.

Studies quantifying response bias may use different methods to obtain information on the non-respondents. First, when access is available to population based registers like health insurance databases, utilization databases or population registers, it is possible to compare respondents with non-respondents with regard to the information provided by the registers. In general, studies using this method [2–4] have shown differences between non-respondents and respondents; for example, non-respondents had lower

annual incomes, more sickness benefit days and were more often unmarried. A drawback of this method is that the response bias can only be examined with regard to the available – often rather general – characteristics in population registers and cannot indicate the degree of response bias regarding the more specific characteristics of interest in comprehensive survey studies.

A second method to quantify response bias is obtaining this specific information by contacting the non-respondents themselves either by telephonic interview or by sending a reply card. A study that used a telephonic interview to obtain information on the non-respondents showed statistically significant differences between respondents and non-respondents for smoking status, hazardous alcohol consumption and lack of vigorous activity [5, 6]. Although such a telephonic interview provides valuable information on non-response, there will always remain a group of non-respondents who either cannot be reached by phone or will be unwilling to participate.

Longitudinal studies provide a third source of information on non-response by allowing the

comparison of respondents and non-respondents at later follow-up, using the information obtained at the start of the study. Most of those studies have found small or no differences between respondents and non-respondents [7–10]. Subjects who repeatedly returned a questionnaire tended to be married, non-smokers and more physically active than those who returned it only once. However, a possible problem with these studies is that they are not based on random samples; the original study population at the first measurement itself may already have been a selected sample.

Here we propose an additional method to obtain information on non-respondents which is based on family and/or twin designs. We will concentrate on general demographic (education), lifestyle variables (smoking, alcohol use, physical activity, coffee- and tea-consumption, religious practice) and personality/mental health (body mass index, burnout, problem behavior, neuroticism). These variables are familial, that is family members resemble each other for those characteristics [11–18]. Therefore, data from non-respondents will be correlated with the data from the respondents and data from responding family members thus will offer information on the non-respondents. We illustrate this approach with data of twins and their siblings collected in 2000 in a survey study on health, personality and lifestyle of the Netherlands Twin Register (NTR).

Acting on the idea that health, lifestyle and personality of the non-responding subjects is reflected by the values obtained on the responding family members we first investigate whether the answers on health, lifestyle and personality variables are different for siblings from highly cooperative families (more than 80% of the family members participated) compared to siblings from less cooperative families (less than 80% of the family members participated). With this method the non-response bias is estimated using information of the responding family members. In addition, we compared data from DZ twins from complete pairs (both twins completed a questionnaire) with data from DZ twins from incomplete twin pairs (co-twin did not participate in the survey study). Data of dizygotic (DZ) twins were used because DZ twins share on average 50% of their genes, just like siblings. However, DZ twins are a select group and may have some distinct features in common which are not generalized to a singleton population (e.g. same age).

Methods

Participants

This study is part of an ongoing twin family study on health-related behavior of the NTR that assesses families with adolescent and (young) adult twins

every two/three years since 1991 [19]. For the present study, the data from the 2000 survey were used.

In May 2000, questionnaires were sent to 13,724 twins/triplets and 2889 siblings. In July 2000 a reminder was sent to the non-respondents. Because smoking was an important theme in the 2000 survey, the reminder contained a pre-stamped reply card with a question on their smoking status (smoker/ex-smoker/non-smoker). The reply card was returned by 2676 persons (2138 twins and 538 siblings) who were not willing to complete the questionnaire. The question on smoking behavior on the reply card was answered by 2473 of the 2676 non-respondents.

Twins and siblings who registered after May 2000 also received a questionnaire ($n = 564$ twins and $n = 776$ siblings) but not a reminder. Twins registered themselves, while most siblings were recruited by asking their mother for their addresses. In total, questionnaires were sent to 14,288 twins/triplets and 3665 siblings from 7223 families. The average family size of the families that were invited to complete a questionnaire was 2.48 (SD 0.99). At the end of the data collection, 4609 twins/triplets and 1474 siblings from 3178 families had completed a questionnaire booklet (Figure 1). For the same sex twins, zygosity was based on questionnaire data or, when available, on DNA typing (zygosity based on DNA was available for 26.1% of the same sex twins). Agreement between zygosity based on questionnaire data and zygosity based on DNA results was 98%. For the opposite sex twin pairs, zygosity is known (DZ) based on their sex. The average family size was 1.91 (SD 0.94). The triplets (41 persons from 22 families), the half-siblings ($n = 27$) and adoption siblings ($n = 5$) were excluded from the analyses.

Data analyses

The percentage smokers were compared for respondents and non-respondents using χ^2 tests.

Familial correlations were calculated for all dependent variables. For the categorical traits tetrachloric correlations were calculated with a threshold model on raw data using MX [20]. The correlations between DZ twins and siblings were constrained to be equal to estimate the familial correlation. For the continuous variables intraclass correlations were calculated from an ANOVA analyses using all DZ twins and sibships [21].

Data from respondents of highly cooperative families were compared with data from respondents of less cooperative families. For each family, the number of respondents was divided by the number of family members who were asked to complete a questionnaire. When less than 80% of the family members participated, the family was marked as 'less cooperative family' and when 80–100% of the family members participated the family was marked as 'highly cooperative family'. The dataset contained

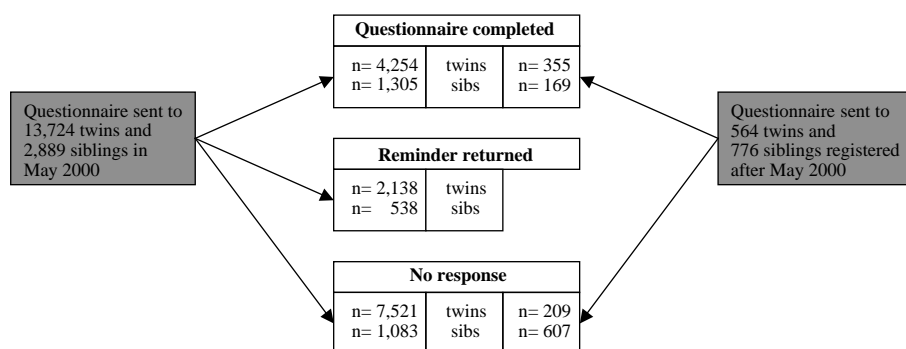


Figure 1. Overview of the number of participants in the study. In May 2000, questionnaires were sent to 13,724 twins and 2889 siblings. A reminder (with reply card enclosed) was sent in July 2000. Subjects who registered after May 2000 also received a questionnaire (twins registered themselves, most siblings were recruited by asking their mother for their addresses). In total, questionnaires were completed by 4609 twins (4254 + 355) and 1474 siblings (1305 + 169). The reply card of the reminder was returned by 2138 twins and 538 siblings who were not willing to complete the questionnaire. Finally, 7730 twins (7521 + 209) and 1690 siblings (1083 + 607) did not return the questionnaire or the reminder (for different reasons such as moved, not interested anymore, died or reason unknown).

1099 families with at least 1 additional sibling. For some of these families more than one additional siblings participated (on average 1.3 sibling per family). From each family one sibling who completed a questionnaire was chosen and scores of the respondents of highly cooperative families ($n = 444$) were compared to the scores of the respondents of less cooperative families ($n = 655$). The mean age of the individuals from highly cooperative families was 30.2 (SD 11.7) and 31.2 (SD 9.8) for the individuals from less cooperative families. Furthermore, 39% of the individuals from incomplete pairs were males and 43% of the individuals from complete pairs were males.

In addition, data from twins from complete DZ twins pairs (both twins completed a questionnaire) were compared with data from twins from incomplete DZ twin pairs (their co-twin did not participate in the questionnaire survey). Data of the monozygotic (MZ) twins were excluded. MZ twins are a special group of siblings who, in contrast to DZ twins or singleton siblings, are genetically identical. When a trait is influenced largely by genetic influences, a MZ twin will be more similar than a DZ twin or singleton sibling. To be able to use the results of this study as example for other large family studies (without twins) we selected the DZ twins and singleton siblings for those analyses. DZ twins share on average 50% of their genes just like singleton siblings, but have, in contrast to other siblings, exactly the same age and are more likely to share similar environmental influences. The DZ twin sample contained 1498 individuals from complete twin pairs and 772 individuals from incomplete twin pairs. The mean age of individuals from incomplete DZ twin pairs was 1 year lower than the mean age of individuals from complete DZ twin pairs (respectively, 28.8 (SD 8.9) and 29.8 (SD 11.4), $p = 0.022$). Furthermore, 33.6% of the individuals from incomplete pairs were males and

32.8% of the individuals from complete pairs were males ($p = 0.339$).

For both comparisons statistical significance was assessed by χ^2 test for categorical variables, by Mann–Whitney test for ordinal variables and by ANOVA for continuous variables. Because multiple comparisons were performed we considered the chance of a type I error. To protect against this error, a Bonferroni correction was used by dividing the significance level by the number of comparisons ($0.05/28 = 0.002$). For a comparison to be considered significant it must have a significance level of 0.002 or less. Statistical analyses were performed using SPSS 11.0 for windows.

Variables

The following variables obtained in the questionnaire were explored in this study:

Smoking

‘Did you ever smoke?’ was recoded to ever smoked (yes) and never smoked (a few times to try, no). ‘How often do you smoke now?’ was recoded to non-smokers (never smoked, never smoked regularly, quitters) vs. smokers (I smoke once a week or less, I smoke more than once a week but not every day, I smoke daily). ‘Do you think you’ll smoke next year?’ (definitely not, probably not, I don’t know, probably yes, definitely yes) was analyzed as numerically ordered variable.

Alcohol consumption

‘Have you ever drunk alcohol?’ was recoded to ever drunk alcohol (yes) vs. never drunk alcohol (a few times to try, no). ‘How often do you drink alcohol?’ was recoded to more than once a week (once a week, several times a week, daily) vs. less than once a week (I do not drink alcohol, once a year or less, a few time

a year, once a month). When at least two questions were answered with 'yes' on the CAGE (a four item questionnaire to detect alcohol problems) the person was classified as possibly having alcohol problems (yes, no).

Coffee- and tea-use

How many cups of coffee/tea do you drink a day? (number of cups/day).

Physical activity

'Do you participate in sports regularly?' (no, yes) and 'Do you cycle regularly?' (no, yes). 'During the last 6 months, how often have you been physical active for more than 20 min?' (never, less than once a month, once a month, 2–3 times a month, 1–2 times a week, 3 times a week or more often) was analyzed as numerically ordered variable.

Religion

'Are you an active member of a religious communion?' (yes I am an active member, I am religious but not a member of a religious communion, no I am not religious) was recoded to 'religious practice' (yes for active members vs. no for non-members and not religious persons).

Burnout

A log transformation was used on the scores of the Dutch version of a five-item subscale (emotional exhaustion) of the Maslach Burnout Inventory-General Survey [22].

Body mass index

A combined measure of height and weight: weight in kg/(height in m)².

Education

Subjects were divided in three groups: low, medium and high education.

Personality

The subscales neuroticism, somatic anxiety, test attitude and extraversion subscales from the Amsterdamse Biografische Vragenlijst (ABV) [23], and the subscales anxious depression, withdrawn, somatic complaints, thought problems, attention problems,

intrusive behavior, aggressive behavior, rule breaking behavior of the Young Adult Self Report (YASR) [24] translated and validated for the Dutch population by Verhulst et al. [25].

Results

The overall response rate for the 2000 survey was 32.3% for the twins and 40.2% for the siblings. Our database consisted of Dutch twin families which were recruited in different ways; a large part of the twin families were recruited by asking city councils in the Netherlands for addresses of twin families, while other twins volunteered to register throughout the study period. The response rates for the survey varied across various subsets of twins. Newly registered twins who volunteered to register and twins who have participated in other waves were more likely to complete a questionnaire. It is important to note that addresses in our database were most up to date for those groups.

Estimating the non-response bias for smoking using a reply card to the non-respondents

To investigate whether a response bias occurred for smoking, the smoking status of the non-respondents was compared with the smoking status of the respondents. Smoking status was available for 6016 respondents (4566 twins and 1450 siblings), as answers on smoking behavior were missing or contradictory for 43 twins and 24 siblings. Smoking status was also available for 2473 non-respondents (1971 twins and 502 siblings) who returned the reply card. As shown in Table 1, the percentage smokers was higher in the non-respondents groups compared to the respondents-groups. A χ^2 test showed that those differences were significant ($p = 0.000$ for men and $p = 0.016$ for women).

Familial correlations for mental health and lifestyle variables

To explore to what extent the variables are familial, the intraclass correlations (continuous data) and tetrachloric correlations (categorical data) were cal-

Table 1. Number and percentage smokers for non-respondents of the 2000 survey (2138 twins and 538 siblings) and respondents of the 2000 survey (smoking status was known for 4566 twins and 1450 siblings)

	Twins			Siblings		
	N smokers	N total	% smokers	N smokers	N total	% smokers
Male respondents	445	1505	29.6	148	580	25.5
Male non-respondents	377	995	37.9	90	268	33.6
Female respondents	730	3061	23.8	196	870	22.5
Female non-respondents	256	954	26.8	61	233	26.2

culated. The lifestyle variables like smoking, alcohol consumption, physical activity and coffee- and tea-use showed familial correlation ranging from 0.21 to 0.44. A high familial correlation was found for religious practice (0.77). The personality and mental health variables showed a somewhat lower familial correlation ranging from 0.11 to 0.20 (Table 2). All correlations were statistically significant.

Estimating the non-response bias using information of the co-twin and siblings

The scores of 655 siblings from a less cooperative family (less than 80% of the family members participated) were compared with scores of 444 siblings from a highly cooperative family (80–100% of the family members participated). The scores for lifestyle variables like smoking, alcohol consumption, physical activity, coffee- and tea-use and personality scores seemed more unfavorable for individuals from less cooperative families, but after Bonferroni correction the differences were not significant (Table 3).

The DZ twin sample consisted of 772 individuals from incomplete pairs and 1,498 individuals from complete twin pairs. The scores of the incomplete DZ twins were somewhat more unfavorable than the

scores of the complete DZ-twin pairs (Table 3). Individuals from incomplete twin pairs cycled significantly less often than individuals from complete twin pairs. They also had significantly higher scores for anxious depression, somatic complaints and neuroticism. The other variables (smoking behavior, alcohol consumption, physical activity, coffee- and tea-use, burnout score, body mass index and education) showed the same trend, namely a more unfavourable score for the individuals from incomplete pairs compared to individuals from complete pairs, but differences were not significant (after Bonferroni correction).

Discussion

The goal of this paper was to examine an alternative method for determining response bias in a survey on health, personality and lifestyle, using data from responding family members as a proxy for non-responding family members. Response rates are dependent on a large variety of factors, such as a monetary incentive, short questionnaires, personalized questionnaires or letters, stamped return envelopes, contacting persons before sending the

Table 2. Familial correlation between DZ twins and singleton siblings

	Familial correlation	<i>p</i> -Value
Ever smoked (yes/no)	0.44	0.000
Current smoker (yes/no)	0.33	0.000
Smoke next year? (five categories)	0.26	0.000
Ever tried alcohol (yes/no)	0.39	0.000
Regular alcohol use: >than once a week (yes/no)	0.33	0.000
Possible alcohol problems CAGE (yes/no)	0.21	0.000
Coffee-use (mean n of cups a day)	0.38	0.000
Tea-use (mean n of cups a day)	0.25	0.000
Regular sports participation (yes/no)	0.25	0.000
Regular cycling (yes/no)	0.21	0.000
Physical activity (six categories)	0.13	0.000
Body mass index	0.38	0.000
Education (three categories)	0.45	0.000
Actively religious (yes/no)	0.77	0.000
Burnout	0.17	0.000
N life events in past 5 years	0.26	0.000
Anxious depression	0.20	0.000
Withdrawn	0.12	0.000
Somatic complaints	0.16	0.000
Thought problems	0.11	0.000
Attention problems	0.15	0.000
Intrusive	0.11	0.000
Aggressive behavior	0.20	0.000
Rule breaking behavior	0.12	0.000
Neuroticism	0.19	0.000
Somatic anxiety	0.18	0.000
Test attitude	0.18	0.000
Extraversion	0.13	0.000

For the categorical variables the tetrachoric/polychoric correlations are calculated. For the continuous data the intraclass correlations are presented.

Table 3. Comparison between individuals from less-cooperative families (<80% of the family members that were asked to complete a questionnaire participated) and highly cooperative families (80–100% of the family members that were asked to complete a questionnaire participated) and comparison between twins from incomplete DZ twin pairs (co-twin did not complete a questionnaire) and DZ twins from complete twin pairs (both twins completed a questionnaire)

	Siblings			DZ-twins		
	<i>L</i>	<i>H</i>	<i>p</i> -Value	<i>I</i>	<i>C</i>	<i>p</i> -Value
% Ever smoked	46.2	42.4	0.222	48.2	44.5	0.096
% Smoker	24.8	22.1	0.313	30.7	27.2	0.081
Smoke next year?	—	—	0.717	—	—	0.048
% Ever tried alcohol	92.6	90.7	0.257	92.3	90.0	0.066
% Regular alcohol use (>than once a week)	42.2	40.7	0.638	37.1	43.2	0.005
% More than once been drunk	56.0	52.2	0.221	55.6	51.0	0.041
% Alcohol problems (CAGE)	12.7	8.6	0.034	9.7	9.5	0.850
Coffee-use (mean n of cups a day)	3.2	2.9	0.323	2.6	2.8	0.090
Tea-use (mean n of cups a day)	2.7	2.6	0.104	2.6	2.4	0.108
% Sports participation	58.3	60.8	0.408	57.4	59.2	0.399
% Regular cycling	63.0	65.8	0.351	57.0	65.9	0.000
How often >20 min physical active?	—	—	0.131	—	—	0.240
Body mass index	24.0	23.6	0.080	22.9	23.0	0.677
Education	—	—	0.455	—	—	0.516
% Religious (active)	24.2	24.0	0.942	18.3	23.6	0.004
Burnout (mean score)	93.7	94.6	0.549	94.5	92.3	0.056
<i>N</i> life events in past 5 years	1.11	1.01	0.471	1.40	1.27	0.011
Anxious depression (mean score)	5.5	5.0	0.057	5.9	5.1	0.000
Withdrawn (mean score)	2.9	2.6	0.091	2.7	2.6	0.049
Somatic complaints (mean score)	3.1	2.8	0.092	3.1	2.7	0.000
Thought problems (mean score)	0.58	0.51	0.987	0.58	0.48	0.097
Attention problems (mean score)	4.3	4.0	0.081	4.5	4.1	0.004
Intrusive (mean score)	3.0	2.7	0.024	2.8	2.7	0.178
Aggressive behavior (mean score)	6.0	5.5	0.038	6.0	5.6	0.039
Rule breaking behavior (mean score)	3.4	3.4	0.933	3.4	3.1	0.063
Neuroticism (mean score)	49.0	45.9	0.038	52.0	47.7	0.000
Somatic anxiety (mean score)	18.1	17.1	0.002	18.1	17.5	0.024
Test attitude (mean score)	37.0	37.5	0.376	37.1	37.9	0.045
Extraversion (mean score)	58.4	58.7	0.708	60.4	60.0	0.606

I = values of the respondents from incomplete twin pairs, *C* = values of the respondents from complete twin pairs. *L* = values of the respondents from less cooperative families, *H* = values of the respondents from highly cooperative families. Comparisons are significant if $p < 0.002$ (Bonferroni correction).

questionnaire and providing non-respondents with a second copy of the questionnaire [26].

The overall response rate of 33.9% in our study may underestimate the actual response rate, as questionnaires were sent to everyone in our database, regardless of earlier participation. Probably, a substantial percentage of the non-respondents are moved to another address. At present, a study is carried out on a sample of non-respondents of a next survey (sent in November 2002/March 2003) which will illustrate the percentage of non-respondents who moved to another address. The exact number of non-respondents due to change in address can best be determined by linking our address database to governmental address databases. However, at the time of writing, Dutch legislation does not allow for use of the official population register to check and update the addresses of our database. Stang [27] concluded that we should not uncritically use the response proportion as an indicator of the likelihood of non-response bias

because there is not always a connection between low response proportions and non-response bias. It is more important to investigate the severity of response bias. Response bias can be explored by different methods like (1) comparison to available data in population based registers, (2) directly contacting the non-respondents by telephone or single-item reply cards, and (3) longitudinal repetition of the survey.

For smoking, we used the second strategy by sending a pre-stamped reply card with a single question on current smoking status to our non-respondents. The results showed a significantly higher percentage smokers among the non-respondents compared to the respondent for both men and women. In most other studies current smoking was also more prevalent among non-respondents [6, 9, 28] or late respondents [29]. The disadvantage of sending a reply card is that only a few questions can be asked. Furthermore, there is a group of non-respondents who neither respond to the invita-

tion to complete the survey nor returns the reply card.

The data collected in twins and their singleton siblings offered a unique opportunity to estimate lifestyle and mental health of non-respondents by the values of their responding family members. Correlations showed that the variables on lifestyle, personality and mental health are familial, that is family members resemble each other for those characteristics. Correlations were higher for lifestyle variables (like smoking, alcohol consumption and physical activity) than for the personality and mental health variables. The higher the familial correlation the better the value of the responding family members may be used as a proxy for the non-responding family members. However, all correlations were significant, and represent the lower bound of the estimates as correlations between same-sex siblings and between siblings close in age are expected to be even higher.

The values of singleton siblings from less cooperative families seemed somewhat more unfavorable than the values of singleton siblings from highly cooperative families although those differences were not statistically significant after Bonferroni correction.

Data on DZ twins are of special interest. Those twins share on average 50% of their genes just like singleton siblings, but have, in contrast to other siblings, exactly the same age. Results showed that individuals from incomplete DZ twin pairs have more unfavorable scores than individuals from complete DZ twin pairs which suggests that the non-responding co-twin is also likely to have an unfavorable lifestyle or lower mental health.

For smoking, both the results of the reply card and the results of the comparison between complete/incomplete twin pairs or highly/less cooperative families suggested a higher percentage smokers among the non-respondents. The differences found with the reply card were statistically significant while the results of the other method were not. Results on non-response bias are probably most trustworthy when collecting data of the non-respondents themselves (by telephone interview or reply card). A limitation of the reply-card approach is the lack of information on completely non-cooperative subjects (subjects who did not respond to the invitation to complete the survey but also did not respond to additional requests for information). The approach to obtain information on non-respondents using the values of their responding family members offers additional information.

In conclusion, the specific composition of our database with twins and their singleton siblings, offered a unique opportunity to estimate lifestyle and mental health of non-respondents by the values of their responding family members. In general, results showed the scores of members from less cooperative families or incomplete twin pairs tended to be lower

than scores from highly cooperative families or complete twins but differences were small. These results suggest that, even for studies with moderate response rates, data collected on health, personality and lifestyle are only mildly biased.

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Address for correspondence: Jacqueline M. Vink, Department of Biological Psychology, Van der Boerhorststraat 1, 1081 BT Amsterdam, The Netherlands
 Phone: +31-20-444-8787; Fax: +31-20-444-8832
 E-mail: jm.vink@psy.vu.nl